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Diagnosis of Sjögren's syndrome via biopsy of the tongue but not the lip: An intriguing case report

Sjögren's syndrome is a chronic systemic autoimmune disease that primarily affects the salivary and lacrimal glands, causing ocular and oral dryness.[1] The diagnosis of Sjögren's syndrome typically involves a biopsy of labial salivary gland. To the best of my knowledge, this is the first reported case of diagnosing Sjögren's syndrome through a biopsy of the tongue but not the lip.

A 62-year-old female patient presented with pain and a foreign body sensation on the left side of the tongue. The patient also complained of sensation of sand or gravel in the eye and pain in the left auricular area. On visual inspection, no specific abnormal findings were observed in the left side of the tongue (Fig. 1A). However, an enhanced CT revealed an enhancing lesion in the left side of the tongue, making it difficult to rule out the possibility of a tumor (Fig. 1B). Also, calcified foci were observed within the left parotid gland (Fig. 1C). An excisional biopsy was performed on the left side of the tongue. On the low-power view, multiple foci of lymphocytic aggregates were observed within the muscle layer of the tongue (Fig. 1D). On high-power view, a severe lymphocytic infiltrate was observed in the salivary gland tissue. Most of the acinar units were destroyed, with only a few ductal structures persisting (Fig. 1E). Notably, the remaining ducts exhibited proliferation and were surrounded by a severe lymphocytic infiltrate, forming the lymphoepithelial lesions (Fig. 1F).

The blood test results showed positive findings, with a rheumatoid factor level of 41 IU/mL (negative if $\leq$ 15), an anti-SSA/Ro antibody level of 2.40 (Index; negative if $\leq$ 0.9), and an anti-SSB/La antibody level of 3.09 (Index; negative if $\leq$ 0.9). In the salivary secretion test, the unstimulated salivary flow rate was 0.464 mL/min, and the stimulated salivary flow rate was 0.894 mL/min, both of which were within the normal range.

The focal lymphocytic sialadenitis in the salivary glands with a focus score (i.e., one or more foci of 50 or more cells per 4-mm^2 area of glandular tissue) greater than 1 corresponded to a score of 3, and anti-SSA/Ro positivity also corresponded to a score of 3, resulting in a total score exceeding 4. Furthermore, sensation of sand or gravel in the eye met the inclusion criteria, and no exclusionary conditions such as sarcoidosis or amyloidosis were present. Based on the American College of Rheumatology/European League Against Rheumatism (ACR-EULAR) classification criteria,[2] a diagnosis of Sjögren's syndrome was confirmed.

Recently, the observation of small, round calcifications in the parenchyma of the parotid gland has gained attention as an effective indicator of Sjögren's syndrome,[3] which closely aligns with the findings in our case (Fig. 1C).

The ACR-EULAR classification criteria designates the lip as the biopsy site,[2] as it is easy to access and less likely to cause excessive bleeding, making it the commonly chosen location.[4] In our case, However, due to the patient's chief complaint and the pathological findings observed in the tongue on CT, a biopsy was performed.

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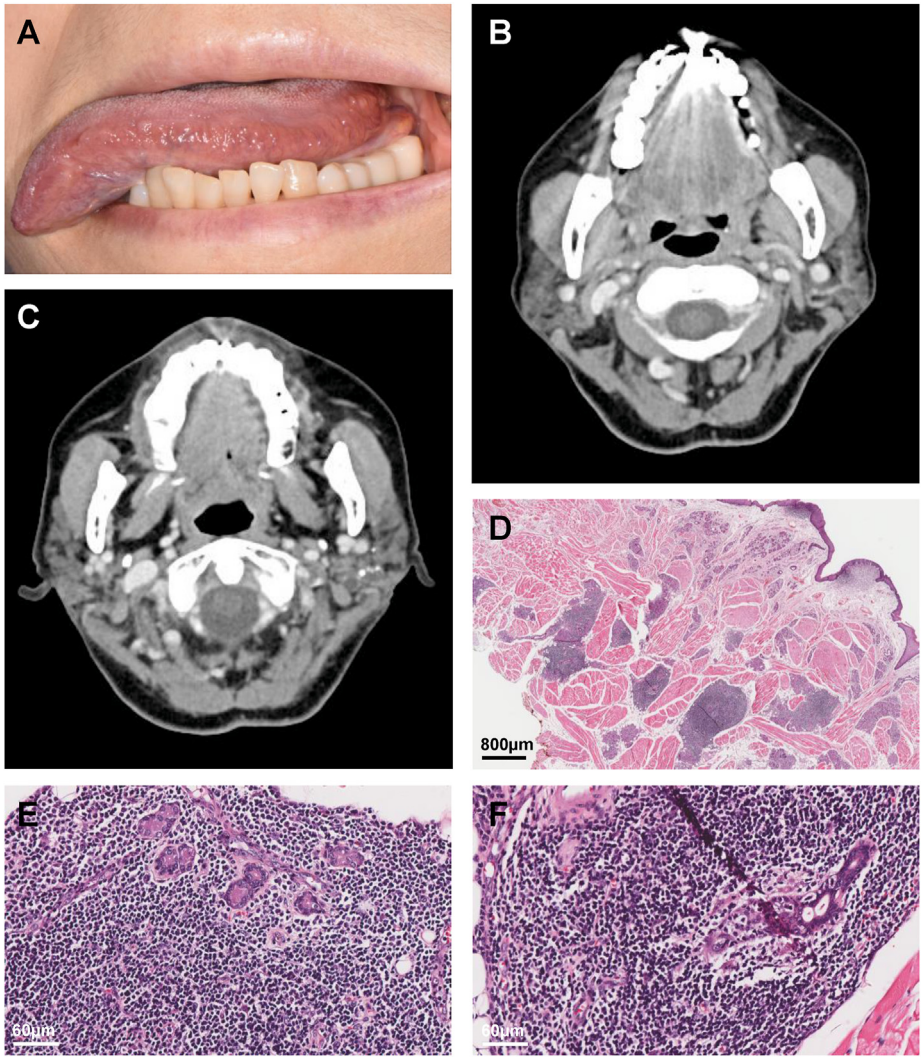

Figure 1 Clinical, radiological and histopathological (hematoxylin and eosin staining) images of the Sjögren's syndrome patient. (A) Clinical photograph of the patient's left tongue showing no specific pathological findings. (B) Enhanced CT image showing an enhancing lesion on the left lateral border of the tongue. (C) Enhanced CT image revealing small, round calcified foci within the left parotid gland. (D) Low-power view showing multiple foci of lymphocytic aggregates among the muscle bundles of the tongue. (E) High-power view showing the lymphocytic sialadenitis, and (F) the lymphoepithelial lesion.

Declaration of competing interest

The author has no conflicts of interest relevant to this article.

Acknowledgments

None.

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